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Abstract

Background/Purpose

Children with type 1 osteogenesis imperfecta (OI) present with abnormal gait characteristics, including reduced power generation during pushoff. However, the exact biomechanical factors associated with reduced power generation are not clearly understood. The purpose of this study was to investigate the biomechanical factors associated with a reduction in ankle power generation in children with type 1 OI.

Methods

Twenty-four participants with type 1 OI (12.5 ± 3.6 years of age) and 24 typically developing children (12.4 ± 3.7 years of age) were evaluated. Three-dimensional gait analysis, isometric plantar flexion strength using dynamometry, and pedobarography were collected on each participant. Results were statistically compared between the groups and a correlation matrix analyzed the associations among the measures.

Results

Children with OI presented with weaker plantar flexors, reduced ankle power generation, and decreased sagittal plane ankle angular velocity during pushoff. However, they presented with similar moment arm distances and ground reaction force magnitudes as typically seen in developing children. There was a higher incidence of pes valgus, increased subarch angles, increased time spent loading the midfoot, and deceased time spent loading the forefoot in children with OI. Plantar flexion strength and the time spent at the midfoot and forefoot were most associated with ankle power generation.

Conclusion

The presence of pes valgus alone does not indicate a reduction of push-off power in children with type 1 OI, but those individuals who have both a flat foot and reduced time spent loading the forefoot during pushoff are the most likely to have reduced push-off power.

Key words

Gait, kinetics, osteogenesis imperfecta, pedobarography, pes valgus

1. Introduction

Osteogenesis imperfecta (OI) describes a group of diseases affecting the bones and connective tissues. It affects approximately one in 20,000 people, with the most common forms linked to mutations in procollagen (i.e., COL1A1 and COL1A2).[1] Type 1 is the most common and mildest form of OI. Individuals have normal quality but reduced quantity of collagen production. Features include autosomal dominant inheritance, bone fragility (prone to fractures), blue sclerae, slightly reduced stature, ligamentous laxity, and muscle weakness.[2] Nearly all children with type 1 OI are ambulatory, and 85% of them walk independently without an assistive device for at least community distances.[3]

Children with type 1 OI present with abnormal gait characteristics, including increased double support time, delayed footoff, reduced ankle range of motion in plantarflexion during preswing, greater ankle power absorption during terminal stance, and reduced power generation during pushoff.[4] These deviations can affect both household and community mobility, but the exact biomechanical factors associated with these abnormalities are not clearly understood.

Ankle push-off power is responsible for approximately 80% of the forward propulsion during normal human walking, and it is often considered to be a major indicator of a mature, efficient, and healthy gait pattern. In the sagittal plane, ankle power generation is calculated by multiplying the ankle flexion moment by the ankle

angular velocity. The ankle flexion moment can be further broken down into the ground reaction force (GRF) value and the moment arm distance. Thus, a reduction in power can result from a decrease in the magnitude of the ankle moment, the angular velocity of the ankle joint, or both.

Another source of reduced push-off power can be weakness of the ankle plantar flexor muscles. In other pediatric populations with known plantar flexor weakness, i.e., cerebral palsy, average peak power was reduced by more than 40%, with a moderate and consistent association between ankle plantar flexor (PF) strength and peak ankle power generation.[5] This same association has never been evaluated in children with type 1 OI. However, previous work has identified that children with type 1 OI present with isometric and functional PF weakness. This weakness was also associated with lower scores in measures of functional mobility.[6]

An additional explanation for the reduction in push-off power in children with OI is that ligament laxity can lead to skeletal malalignment under certain loading conditions. Collapsing into pes valgus during a prolonged transition from midfoot to forefoot loading creates a biomechanical disadvantage for the foot to transmit a force with the necessary magnitude and direction for typical power generation. This condition is commonly referred to as “flexible lever arm dysfunction,” and it is comparable with prying a heavy load with a rubber crowbar.[7]

Pedobarography is a useful clinical tool that quantifies dynamic foot function and helps to examine the foot for abnormal mechanics. A pedobarographic technique has been recently developed and validated to help identify the presence of flexible lever arm dysfunction.[8] This technique identifies the medial-lateral location of the center of pressure progression (COPP), which is the relative movement of the center of pressure on the plantar surface of the foot during the stance phase of gait. This medial-lateral path can then be used to identify the presence of varus-valgus at the hindfoot (HF), midfoot (MF), forefoot (FF), and/or the entire foot. It also measures the percentage of stance phase spent loading each segment of the foot. The location and timing of the COPP, along with foot geometry measures (i.e., the longitudinal arch angle), can help detect the presence of flexible lever arm dysfunction during locomotion in children with type 1 OI. This information can then be used to identify any associations between COPP location and reduction in push-off power. These methods of pedobarographic analysis have not been used to study children with OI, but the analysis of peak pressure and the pressure time integral have been associated with kinetic gait abnormalities in patients with diabetic neuropathy.[9]

1.1. Aim

The purpose of this study was to investigate the biomechanical factors associated with reduction in push-off power in children with type 1 OI. We analyzed gait kinematics and kinetics, along with isometric strength and pedobarographs in typically developing children and children with type 1 OI to address the following hypotheses: (1) children with type 1 OI have a shortened ankle moment arm and reduced magnitude in the GRF at the time of peak ankle flexion moment (terminal stance) compared with typically developing children, (2) children with type 1 OI demonstrate a higher incidence of pes valgus and flatter feet than typically developing children, and (3) push-off power is correlated to measures of flexible lever arm dysfunction and isometric PF strength.

2. Methods

2.1. Participants

Twenty-four participants with type 1 OI (“OI Group”, 11 males, 13 females; 12.5 ± 3.6 years of age) and 24 age and sex-matched typically developing children (“Control Group” 12.4 ± 3.7 years of age) gave their informed consent to participate in this Health Insurance Portability and Accountability compliant, Institutional Review Board (Shriners Hospitals for Children, Chicago, IL, USA, and RUSH University, Chicago, IL, USA)-approved

protocol. All participants in the study were community ambulators without assistive devices who reported no pain on the day of testing. Three participants were being treated with intravenous bisphosphonate therapy and one was on an oral bisphosphonate regimen at the time of testing. No participants had experienced a fracture or surgery 1 year prior to this investigation, although one participant had a fracture within 2 years. Two participants had a surgical intervention that consisted of an epiphysiodesis for a leg-length discrepancy and intramedullary rodding of the femur 3 years prior to participating in this study.

2.2. Protocol

Each participant underwent a lower-extremity strength assessment as well as a gait analysis that included kinematic, kinetic, and dynamic pedobarography while walking at a self-selected speed.

2.3. Strength assessment

Isometric strength testing of the ankle PFs and dorsiflexors was completed on the Biodex System 3 dynamometer (Biodex Medical Systems, Inc., Shirley, NY, USA). Participants were seated with the thigh posteriorly supported, placing the hip in a flexed position and the tibia parallel with the floor in accordance with the instruction manual. The average peak torque of the three trials was normalized to body weight and was represented as a percentage of total body weight.

2.4. Gait analysis

Kinematic and kinetic data were collected with a 14-camera Vicon MX Motion Analysis System (Vicon Motion Systems, Inc., Lake Forest, CA, USA) synchronized with four 6-DOF force plates (AMTI, Waterton, MA, USA) embedded in the laboratory walkway. Kinematic (video) data were collected at 120 Hz, and kinetic (force plate) data were collected at 1,080 Hz. Three dimensional joint kinematics and kinetics were calculated using the PlugInGait model.[10] All joint moments were calculated using inverse dynamics and reported as internal (demand) moments. Walking speed was also calculated. Moment arm values were calculated by measuring the distance of the ground reaction force to the ankle joint center. Ankle joint angular velocity was calculated by taking the slope of the kinematic plot during one representative gait cycle.

2.5. Dynamic pedobarography

Pedobarographic data were collected at 50 Hz using an EMED ST2 pedobarograph (Novel, Munich, Germany) with a sensor area of 38 by 72 cm and a sensor density of 4 sensors/cm^2. Three representative trials for each foot were averaged together. The location and duration of the COPP was tracked through each segment of the foot (HF, MF, and FF) during stance phase.[8] The medial longitudinal arch angle was calculated using the geometry program (Novel, Munich, Germany) from the maximum pressure picture of the foot (Figure 1).

Figure 1. The subarch angle is defined as taking the point of the most lateral displacement of the border of the medial midfoot and drawing a line from that point to a point tangent to the border of the medial forefoot segment and a line tangent to the border of the medial hindfoot segment.

2.6. Statistical analysis

For all tests, three representative trials were averaged together for each group (OI and control). Descriptive statistics were calculated for all interval variables and were analyzed using a Student's t-test with significance set at $p < 0.05$. Pearson Correlation Coefficients were also calculated for all interval variables to examine the various gait, strength, and pedobarographic data's relationship to peak ankle power generation. For all correlations, an r of 0.75 to 1.00 was considered good to excellent, 0.50 to 0.75 was considered moderate to good, 0.25 to 0.50 was fair, and 0.00 to 0.25 indicated little to no relationship.[11]

3. Results

Gait analysis confirmed several parameters were reduced in the participants of the OI group, including peak ankle power generation, peak ankle flexion moment, walking speed, and peak ankle plantarflexion at pushoff. Measures that were not significantly reduced were maximum moment arm distance and maximum GRF compared with the control group (Table 1).

Table 1. Gait data and isometric strength group averages

Gait measures	**OI group**	**Control group**
Peak ankle power generation (W/kg)	2.7*	3.7
Peak ankle flexion moment during gait (Nm/kg)	1.0*	1.2
Maximum moment arm (mm)	124.5	125.4
Maximum ground reaction force (N)	10.8	11.3
Peak ankle plantarflexion angular velocity (°/sec)	316.4*	382.2
Walking speed (m/s)	0.95*	1.16
Peak plantarflexion during preswing (degrees)	10.7*	15.6
Peak plantarflexion strength on dynamometer (Nm/kg)	79.8*	101.7

OI = osteogenesis imperfecta.

∗Indicates significantly different than the control group ($p < 0.05$).

Isometric strength measurements were compared and revealed significant weakness for Peak Ankle Plantar Flexion in the OI Group compared with the control group (Table 1). A small amount of the absolute power was used during gait. The strength required to generate the peak ankle flexion moment during gait is only 1.25% of the peak plantar flexion strength generated on the dynamometer during testing for max strength in the OI group and 1.18% of the max strength for the control group.

Pedobarography data describing the duration of the COPP location in the HF, MF, and FF segments during stance revealed reduced loading time of the OI group's FF and increased loading time of the HF and MF compared with the control group (Figure 2). Segmentally, valgus was observed throughout the foot with the MF > HF > FF (Table 2) in the OI group. The subarch angle was 32% greater in the OI group, which indicated that more of the MF is in contact with the plantar surface (Table 2).

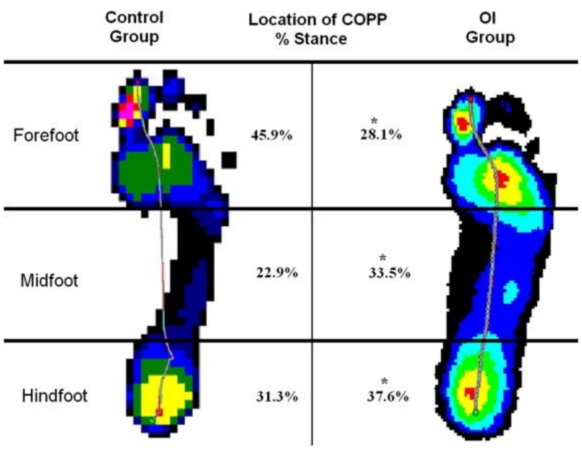

Figure 2. Pedobarography analysis of the center of pressure progression (COPP) location during stance phase. Each group's feet are flipped and shown as right feet, then averaged together. *Indicates significantly different than the control group ($p < 0.05$).

Table 2. Pedobarography data group averages

Pedobarography measures	OI group	Control group
Longitudinal arch angle (degrees)	136.9*	104.0
OI group with HF valgus	38%	—
OI group with MF valgus	52%	—
OI group with FF valgus	14%	—

FF = forefoot; HF = hindfoot; MF = midfoot; OI = osteogenesis imperfecta.
∗Indicates significantly different than the control group ($p < 0.05$).

The correlation matrix in Table 3 displays the associations among the measures of foot biomechanics and gait parameters. Excellent correlations were observed between the max power generation and the amount of time spent loading the FF. Moderate correlations were seen among isometric PF strength, max power generation, and COPP at the FF. A moderately negative correlation was observed between the COPP at the MF and the max power generation as well as isometric PF strength and the COPP at the MF.

Table 3. Osteogenesis imperfecta group correlation matrix

	Max Power generation	Subarch angle	COPP time at MF	COPP time at FF	Isometric PF strength
Max power generation	—	−0.29	−0.31*	0.75*	0.57*
Subarch angle	−0.29	—	0.45*	−0.45*	−0.31
COPP time at MF	−0.31*	0.45*	—	−0.42*	−0.12
COPP time at FF	0.75*	−0.45*	−0.42*	—	0.60*
Isometric PF strength	0.57*	−0.31	−0.12	0.60*	—

*$p < 0.05$.
COPP = center of pressure progression; FF = forefoot; MF = midfoot; PF = plantar flexor.

4. Discussion

This study aimed to determine what biomechanical factors are associated with reduced peak ankle power generation (i.e., pushoff) during gait in a group of 24 participants with type 1 OI. Gait analysis, strength, and

pedobarographic data were used to uncover the underlying source of the reduction in push-off power. Previous research has identified several gait deviations using quantitative gait analysis in this population.[4] Because ankle power generation is generally accepted as one of the key components of a typical, efficient gait pattern, we chose to focus on that measure.

4.1. Kinematic and kinetic differences

The first hypothesis, which stated that participants with type 1 OI are walking with reduced lever arm distances and GRF magnitude, was not supported because we found that participants with type 1 OI were generating a nearly identical moment arm distance from the ankle joint and magnitude of GRF during peak push off compared with those in the control group. This was somewhat surprising because we thought the OI group would use a gait strategy that would reduce high moments and, thus, reduce the torques applied to fragile bones and joints. It is important to note that the percentage of stance phase spent with the COPP within each segment of the foot was different between groups. Specifically, increased stance phase times were observed in the OI group at the HF and MF segments, with decreases in the FF segment.

Additionally, reductions were found in the ankle joint angular velocity, specifically the rate at which the OI group plantar flexed the joint; however, this may be attributed to multiple parameters. One of those parameters could be because the OI group had a reduced walking speed; in elderly populations, research suggests that walking at slower speeds can be a contributing factor to lower ankle power generation.[12] Also, as previously stated, the OI group spent an increased amount of time loading the MF and a decreased time loading the FF. Individuals with reduced FF loading time and increased time loading the MF typically generated less ankle power at pushoff. This is consistent with the theory of lever arm dysfunction where prolonged loading of the MF segment results in deformation instead of joint rotation.

Strength data analysis did reveal PF weakness in the OI group compared with the control group; however, the strength needed to generate the torques at the ankle during gait are well within the capabilities of the participants in the OI group. Therefore, their level of strength may not be a functional deficit in terms of gait.

4.2. Pes valgus

Analysis of the pedobarographs supported the second hypothesis, i.e., that the participants with OI had flatter feet with a collapsed medial/longitudinal arch and valgus COPP throughout the foot. These data fit the pathology associated with OI in which the reduction in the quantity of collagen results in ligamentous laxity. Pes valgus has been previously cited as a contributor to lever arm dysfunction that results in a loss of ankle power generation.[7]

4.3. Flexible lever arm dysfunction

The third hypothesis attempted to link measures of pes valgus with the reduction in push-off power to further support the theory of flexible lever arm dysfunction. However, measures of the pes valgus, such as the subarch angle, did not demonstrate a significant correlation with push-off power generation. Significant correlations were instead supported by COPP measures (i.e., the amount of time spent loading the segments of the foot). This leads to the interpretation that observing the time spent loading the segments of the foot provides a greater insight into ankle power generation than the amount of pes valgus. Further support for analyzing the timing of peak events during gait is supported by previous research of sagittal plane ankle kinematics in children with type 1 OI.[4] These data showed that there was an increase in the peak ankle dorsiflexion leading to a prolonged midstance phase (second rocker) of the gait cycle in individuals with type 1 OI. This corresponds with a reduction in the amount of ankle joint plantar flexion and shortened preswing phase (third rocker).

Biomechanically, prolonged loading of the midfoot during single limb support as seen in the gait pattern of individuals with type 1 OI may be an indication of the unlocking and collapse of the midtarsal joints, the

excessive stretching of foot ligaments due to laxity, or both. This laxity prohibits the foot from becoming a rigid lever arm capable of transmitting the full magnitude of the propulsive force. By the time the foot has completely approximated with the floor and become rigid enough to allow for full loading in the metatarsal heads, it is postulated that the shank has already advanced well anterior to the ankle joint center and pushoff is near its end phase with little opportunity to generate adequate ankle power.

A limitation in this study was our inability to test for the avoidance of repeated high forces in the small bones in the foot. It may be that these individual's prior experience with fractures and the inherent fragility of their bones causes them to avoid repeated loading of the FF, as during gait, and, therefore, reduce their ankle push-off power. Further research is necessary to study the material properties of these individuals' bones to assess their biomechanical limitations.

5. Conclusion

Individuals with type 1 OI may have the capability to create the force and moment arm necessary to generate normal ankle power, but multiple temporal loading and biomechanical factors may inhibit power generation. We propose that the presence of pes valgus alone does not indicate a reduction of push-off power, but those individuals who have both a flat foot and reduced time spent loading the forefoot during pushoff are the most likely to have decreased push-off power. Individuals with OI may be using an avoidance strategy in an attempt to minimize the increased loading of compromised foot and ankle structures prone to fracture.

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